Pilot and Feasibility Studies

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Exercise-based telerehabilitation for the management of chronic pain in people with severe haemophilia: a mixed-methods feasibility study

Paul McLaughlin[1,2*], Pratima Chowdary[1,2], Kate Khair[3], Clive Smith[4], David Stephensen[5] and Michael Hurley[6]

Abstract

Background Chronic pain is reported by between 30 and 71% of people with haemophilia (PWH). Exercise is shown to be effective for pain management in other arthritides, but it remains unclear if such an approach is effective or acceptable to PWH. The aim of this study was to evaluate the feasibility and acceptability of a telerehabilitation exercise intervention for PWH living with chronic pain.

Methods This was a multisite, non-randomised, pre-post feasibility design, with a nested qualitative study. People with severe haemophilia > 18 years, living with chronic pain, were recruited. The intervention comprised 12 low-impact/moderate intensity, individualised exercise sessions and 3 knowledge-sharing and discussion sessions. Primary objectives assessed according to predefined progression criteria were as follows: (a) recruitment rate (5 participants enrolled per site over 8 weeks), (b) adherence (≥ 75% participants would adhere to ≥ 75% of sessions), (c) follow-up rate (≥ 75% completion of self-reported measures), (d) fidelity (intervention delivered as described in protocol) and (e) safety (≤ 30% participants would report adverse events). Acceptability was evaluated from thematic analysis of post-intervention participant interviews. Preliminary evaluation of self-reported pain, function and quality of life (QoL) was a secondary objective. Results were reported using descriptive statistics integrated with qualitative findings.

Results Ten PWH were recruited and completed the intervention. Nine agreed to be interviewed post intervention. Attendance at individual sessions was 84.5% compared to 52.1% for the group sessions. Outcome measures were successfully completed for 100% at baseline, 70% at intervention end and 60% at 3-month follow-up. No serious adverse events were recorded. Group median values in outcome measures (pain, function, QoL) showed minimal change post intervention. Participant interviews highlighted high levels of enjoyment, confidence in continuing exercises independently and positive views of virtual delivery and condition-specific exercise.

Conclusions Recruitment rate and safety met the predefined progression criteria. Fidelity partially met the progression criteria, but the follow-up rate for self-reported measures did not. The study was acceptable to both participants and physiotherapists. Further intervention development is needed to review approaches to outcome measure collection and refine the usefulness of the knowledge-sharing sessions.

*Correspondence:
Paul McLaughlin
p.mclaughlin@nhs.net
Full list of author information is available at the end of the article

Trial registration The study was prospectively registered on 9 July 2021: International Standard Randomised Controlled Trial Number ISRCTN 17454597.

Keywords Haemophilia, Pain, Exercise, Rehabilitation, Telerehabilitation, Feasibility

Key messages regarding feasibility

- What uncertainties existed regarding the feasibility?

Chronic pain associated with haemophilia arthropathy is a significant clinical issue. Current haemophilia treatment guidelines recommend a primarily pharmacological approach to chronic pain management with the use and approach of physiotherapy being discrepant. Whilst general exercise is now accepted as safe for PWH, the acceptability and feasibility of an exercise-based approach in managing chronic pain are unknown. Furthermore, there is a dearth of knowledge on the applicability and usefulness of delivering such an intervention using virtual communication technology.

- What are the key feasibility findings?

This study confirmed that a telerehabilitation intervention was feasible and acceptable to PWH living with chronic pain. There were no serious adverse events reported. Whilst the virtual delivery was highly acceptable to participants, the physiotherapists reported an increased administrative burden in delivering the study. Participant-reported outcome measures did not fully capture change experienced by participants, but post-intervention interviews did. Inclusion of knowledges and discussion sessions did not appear to provide added value to the intervention.

- What are the implications of the feasibility findings for the design of the main study?

Virtual delivery was highly regarded by participants in terms of time and convenience, although both participants and physiotherapists felt that having an option of face to face as well as virtual may be beneficial after more than 6 weeks. Further work is needed to establish which outcome measures may be more acceptable to monitor change as well as be meaningful to those taking part. Given the burden on the physiotherapists, further evaluation of feasibility is required for delivery of this study in services with less than a full-time physiotherapist.

Background

Haemophilia is the umbrella term for the most common of the rare lifelong bleeding disorders — haemophilia A (deficiency of clotting factor protein VIII) and haemophilia B (deficiency of clotting factor protein IX). Both disorders occur due to a mutated or absent genetic code on the X chromosome, and therefore, it almost exclusively affects males [1, 2]. Disease severity is based against normal values for clotting factors of 50–150%: mild (levels between 5 and 40% of normal), moderate (between 1 and 4% of normal) and severe (levels of < 1% of normal) [3, 4]. In its untreated state, spontaneous musculoskeletal bleeding is a hallmark of the condition in almost all people with severe and some with moderate haemophilia. The mainstay of current treatment is to raise the factor levels in the blood or balance haemostasis enough so as to limit the possibility of spontaneous bleeding (prophylaxis) or treat if a bleed is suspected (on-demand) [3].

The phenomenon of intra-articular joint bleeding in haemophilia is proposed to initiate the process of synovial joint destruction in three interrelated stages of acute haemarthrosis, synovitis and degenerative joint arthritis [5]. Haemophilic arthropathy (HA) is the term given to this process and is characterised by chronic synovitis, cartilage destruction, epiphyseal enlargement and bony deformity [6] and has been shown to have predominantly degenerative, rather than inflammatory, characteristics [7]. A recent UK study of data from the National Haemophilia Database highlighted that those younger PWH (< 19 years old) had little or no joint damage due to having treatment since infancy, whereas those over 40 years old had significantly higher levels of joint disease [8]. Increasing severity of joint damage alongside the increased number of joints affected in older adults with severe haemophilia has also been shown to be strongly correlated to poor perception of function and moderately correlated with pain [9, 10].

Current data indicates that the experience of pain is an unavoidable reality for many PWH, with figures suggesting between 49 and 61% of PWH experience pain on a daily basis [11, 12]. Episodic acute pain is reported in 20–68% of adults [12, 13] with chronic pain experienced by 30–71% of adults [12, 14] and in 19% of children [15]. Living with haemophilia and chronic pain brings with it constraints in mobility and independence, increased

anxiety, poor quality of life and frustration due to restrictions in activities of daily living [16, 17].

Figures vary from 21 to 50% of PWH reporting that they did not believe their pain was well managed [12, 18] and may reflect a lack of standardised management pathways. Interestingly, physiotherapy as an option in pain management is discrepant, reportedly used by between 12 and 46% of people [19, 20]. However, what that physiotherapy may entail and in what context (acute or chronic pain) is poorly described, as is the effectiveness of such physiotherapy intervention. Whilst effectiveness of rehabilitation for primary management of pain in osteoarthritis and rheumatoid arthritis is well established [21–23], there has been no structured scientific research to evaluate its effectiveness for management of chronic pain in PWH. Whilst exercise in general has been shown to be safe for PWH [24], its use as an option in pain management remains unknown.

Physiotherapy for PWH has traditionally been delivered in person in haemophilia centres. However, even prior to the Covid-19 pandemic, the use of a telemedicine approach was being highlighted as an opportunity for specialist haemophilia care to be delivered locally to those living large distances from their specialist centres [25]. Telemedicine approaches for multiple aspects of haemophilia healthcare delivery have been shown to be acceptable to PWH throughout the Covid pandemic relating mostly to the delivery of routine care, e.g. review appointments over the telephone or on webcam [26]. In relation to physiotherapy specifically, there has been tentative feasibility demonstrated in the virtual delivery of general exercise classes for PWH [27, 28], as well as co-developed hybrid interventions (mix of face to face and virtual) for those living with haemophilic arthropathy [29] and to increase confidence in being physically active [30]. Feldberg and colleagues evaluated the use of an asynchronous exercise and pain education intervention (videos) for chronic pain in PWH, reporting positive improvements in pain intensity and function [31]. However, to date, no intervention has been developed or evaluated that uses a real-time (synchronous) telerehabilitation approach specifically developed for use in the management of chronic joint pain in PWH.

The UK Medical Research Council (MRC) framework defines the importance of feasibility testing in the development of new complex interventions [32]. In considering the level of complexity presented by PWH living with painful haemophilic arthropathy, the design and effective components of a rehabilitation intervention and its potential use for pain management in PWH have not previously been evaluated. Furthermore, the potential feasibility of a telerehabilitation approach for delivering an exercise-based intervention for PWH with chronic pain remains unknown.

The overall aim of this study was to evaluate the feasibility and acceptability of a physiotherapy-led, low-impact, moderate intensity telerehabilitation intervention in PWH who have chronic joint pain related to haemarthropathy, termed the REMAP-Haemophilia study (*RE*habilitation for the *M*anagement of Arthritic Pain in haemophilia). Evaluation of objectives were carried out using quantitative and qualitative approaches.

The primary objectives identified for the study were as follows:

1. Determine the safety of an exercise-based telerehabilitation intervention for PWH.
2. Evaluate the feasibility and acceptability of the intervention delivery and content.
3. Determine the acceptability of the overall intervention (recruitment rate, adherence to the intervention, attrition and study completion rate).
4. Determine the acceptability of chosen outcome measures.

The secondary objective identified for the study was as follows:

1. Collection of preliminary efficacy data (before and after) with patient-reported outcome measures (PROMs) evaluating pain, quality of life and function.

Methods

Study design

This was a multisite, non-randomised, pre-post feasibility study with an explanatory-sequential nested qualitative study.

The study was given ethical approval by the East Midland-Nottingham 2 Research Ethics Committee (rec. reference number: 21/EMI/0161). The study was sponsored by the Royal Free London NHS Foundation Trust (reference number: 141604) and was prospectively registered (ISRCTN 17454597).

Participants

Participants were identified by the physiotherapist in advance of attendance at routine haemophilia clinic reviews. Following eligibility screening, participants were given a study information sheet. Written consent was obtained prior to completion of baseline assessments. Baseline demographic data was collected from the medical notes.

The study inclusion criteria were as follows:

- People with severe haemophilia A or B (with or without an inhibitor)
- Aged 18 years and over
- Self-reported symptoms of chronic pain associated with haemophilic arthropathy in any joint
- Willing and able to give informed consent for participation in this study
- Able to follow instructions
- Have a good command of written and spoken English
- Registered at a UK-located haemophilia comprehensive care centre with a named physiotherapist
- Have access to a laptop/tablet with webcam at home and sufficient Internet connection

The study exclusion criteria were as follows:

- People with mild or moderate haemophilia A or B
- Any other inherited bleeding disorder
- A diagnosis of chronic pain that is not associated with HA
- Severe and/or unstable cardiovascular disease
- Severe and/or unstable pulmonary disease

Intervention

In keeping with the MRC guidelines for the development of complex interventions, REMAP-Haemophilia used stakeholder participation to develop the theory underpinning the intervention. Stakeholders included people with haemophilia, specialist haemophilia physiotherapists and clinical academics with experience in intervention development [33]. Prior qualitative studies also informed aspects of the intervention and outcome measure choice [34, 35]. The theory development process also informed the identification of behavioural change techniques (BCT's) to include in the overall design and delivery of the intervention (the full list of BCTs is provided in Supplementary File 1).

The overall design and delivery of the study are described in Table 1 according to the Template for Intervention Description and Replication (TIDier) and Consensus in Exercise Reporting Template (CERT) checklists [36, 37].

The REMAP-Haemophilia study was a 12-session (6 week), low-impact, moderate intensity exercise-based intervention delivered virtually using the Microsoft Teams digital platform. One individual exercise session and one group exercise session were planned each week, with appointment times agreed between the physiotherapist and participants. An initial face-to-face assessment provided each participant with their starting point for each exercise. Exercises targeted upper and lower limbs, as well as comfortably challenging cardiovascular effort. The lower impact approach aimed to limit mechanical stress on those with haemophilic arthropathy of the ankle.

A physiotherapist led every session, gave the instructions for each activity, monitored effort and participation ability, provided feedback and encouragement and kept time. Each exercise was repeated three times per set and timed at 30 s of moderate exertion (as per the rated perceived exertion score card that each participant had at home), 30-s rest and a 2-min break in between each set. Exercises included resistance (body weight or additional) and cardiovascular with an additional exercise being added to the overall session plan every 2 weeks. The total time needed per exercise session was designed not to exceed a total session time of 40 min by the end of week 6. Participants were not restricted from participating in their normal routines, nor were they prevented from commencing new physical activities whilst taking part in this study (detail of the exercises can be found in the Supplementary File 2).

Three 'knowledge-sharing and discussion' sessions were delivered by the physiotherapist before the group exercise session at weeks 1, 3 and 5. Delivered over MS Teams, the sessions focussed on the following: (1) why we experience pain and what is means, (2) physical activity (benefits and struggles) and (3) pacing and finding your own level. After a short presentation, the aim was to encourage a forum for participants to discuss shared experiences of pain and activity and any actions or activities they had found to be helpful for them (detail of the sessions can be found Supplementary File 3).

Physiotherapists received training in advance of the study commencement which included delivery of protocol, study delivery/management requirements and delivery of the BCTs.

Participant-reported outcome measures

The PROMs were collected at three time points — pre intervention (T0), on intervention completion (T1) and at 12-week post-intervention completion (T2). The pre-intervention measures were collected in person at the initial face-to-face session, with the remaining two time point outcome measures being posted to participants with a prepaid, addressed envelope.

Measures of pain

Brief Pain Inventory (BPI-SF)

This 9-item self-administered questionnaire evaluates the severity of a person's pain and its impact on their daily functioning and is widely used in a range of

Table 1 Exercise intervention summarised as per the CERT and TIDier checklists

Item	Description
Name	*RE*habilitation for the *M*anagement of *A*rthritic *P*ain in Haemophilia — the REMAP-Haemophilia study
Why: Rationale	To evaluate the feasibility and acceptability of an exercise-based telerehabilitation intervention for people with severe haemophilia and chronic pain
What: Materials	• Hardware: Wi-Fi, webcam on computer/tablet/ telephone • Software: Video-conferencing platform (Microsoft Teams) • Equipment: Resistance exercise bands
What: Procedures	○ Embedded behaviour change techniques throughout study design and delivery ○ Exercise prescription plan agreed between participants and physiotherapist ○ Pre-procedure outcome measures completed (T0) Two exercise sessions per week over 6 weeks • Low impact, moderate intensity • Targeting both upper and lower limbs and cardiovascular ○ Three knowledge and discussion sessions • Weeks 1, 3 and 5 ○ Post-participation interviews — PWH and physiotherapists ○ Post-procedure outcome measure on intervention completion (T1) and at 12-week post-completion (T2)
Who: Provider	Specialist haemophilia physiotherapist working in a comprehensive care haemophilia centre, trained in the delivery of the REMAP-Haemophilia protocol
How: Delivery	• 1 × in-person session: Completion of outcome measures and practice exercises with the physiotherapist to find appropriate starting point • 1 × technical dry run with participants of using webcam for exercise when back in their own home • 1 × individual and one group exercise session per week over 6 weeks — real time, virtual delivery • 3 × knowledge and discussion sessions — real time, virtual delivery prior to the group exercise session • Post-participation interviews — telephone or video-conferencing platform
Where: Location	Participants in their own homes and physiotherapists are hospital based
When, how much: Dosage	• Frequency two times per week • Moderate intensity as per the Rated Perceived Exertion Scale — every participant had own copy at home • Duration between 25 and 40 min — increased every 2 weeks as new exercise added to session
Tailoring: What and how	• Each exercise had three starting points depending on individual participant ability and pattern of joint disease and function • Participant starting point practised and agreed with physiotherapist • Advice from physiotherapist on alteration to exercises within sessions if participant experienced difficulty
How well: Planned	• Physiotherapists to keep notes on each session and their own diary • Participants to keep weekly diary about experience and feelings taking part in study • Post-participation interviews with participants and physiotherapists
How well: Actual	Evaluation of both qualitative and quantitative findings to inform feasibility, acceptability, safety, and efficacy of intervention

non-malignant pain conditions [38]. Test–retest reliability construct validity is good when used in PWH [39, 40], but responsiveness is as yet unknown.

Pain Self-Efficacy Questionnaire (PSEQ)

This is a 10-item questionnaire that assesses the confidence of people (with any type of chronic pain) in activity despite pain. Each item's response is on a 7-point scale and is scored 0–6. It is an additive score between 0 and 60, whereby a higher score indicates higher self-efficacy beliefs [41]. Validity, reliability and responsiveness in people with musculoskeletal disorders are excellent [42], but its use has not previously reported in PWH.

Measures of quality of life

EQ5D-5L

This is a 5-item questionnaire evaluating generic health-related quality of life over five dimensions (mobility, self-care, usual activities, pain/discomfort and anxiety/depression). It is reported as an overall utility score, alongside an overall health report with a visual analogue scale [43]. It has been shown to have satisfactory construct validity in PWH [39, 44].

Musculoskeletal Health Questionnaire (MSK-HQ)

This 14-item questionnaire allows people with MSK conditions to report their symptoms with questions relating to pain/stiffness in the day and night, problems with activities of daily living, sleep disturbance, emotional wellbeing and confidence in managing symptoms. It is scored additively from 0 to 56, whereby a higher number indicates better musculoskeletal status. Whilst its use has not previously reported in PWH, it has been shown to be responsive across a range of musculoskeletal conditions [45].

Measures of function

Haemophilia Activities List (HAL) Questionnaire

This measures the impact of haemophilia on self-perceived functional abilities in adults with haemophilia. It has 42 multiple-choice questions across seven domains. The total score ranges between 0 and 100, where a higher score indicates less perceived functional impairment. It has been shown to have good internal consistency and convergent validity in PWH [46].

Patient-specific functional score (PSFS)

This is a self-reported measure that aims to assess functional change in people presenting with predominantly musculoskeletal disorders. Participants identify up to five activities that they are having difficulty with as a result of their problem, rating the current level of difficulty associated with each activity on an 11-point scale (0–10). They then rescore at the end of the intervention.

Measuring overall change

Patient global impression of change (PGIC)

This is a single question completed at the end of the intervention that measures a change in an individual's clinical status. People rate the change in their own clinical status on a 7-point scale, from very much improved to very much worse.

Diaries

Participants completed a short weekly diary reflecting on their experience that week and any change they noticed in themselves. They were asked to send their completed diaries back in the same envelope as the outcome measures at T1.

The physiotherapists delivering the study also completed a weekly diary to record their thoughts and reflections on practicalities of delivery of the study, feedback or comments they had received from participants, technical issues and any changes they made to how they delivered the study.

Qualitative evaluation

All those completing the exercise session component of the study were contacted by email or telephone to confirm if they still wished to be interviewed, with an interview arranged at a time convenient for them.

A topic guide developed with a PWH was used for the post-intervention interviews. Semi-structured interviews were conducted over Microsoft Teams or on the telephone. Questions were open ended and aimed to gain an insight into each person's experience of taking part in the exercise intervention, as well as drawing focus to the objectives relating to the feasibility and acceptability of the intervention.

Each participating physiotherapist was interviewed over MS Teams on completion of the intervention with a focus on feasibility, acceptability and fidelity of the delivery of the study, as well as general feedback and overall views of having taken part.

All interviews were recorded, and the audio files transcribed verbatim by a professional transcription service.

Feasibility outcomes and progression criteria

Safety of the intervention was evaluated by the number of reported adverse events/serious adverse events recorded at each site. Perception of safety was also evaluated in the participant post-intervention interviews.

Progression criteria for feasibility outcomes were identified a priori and are detailed in Table 2. Failure of an outcome to meet progression criteria would be evaluated against the need for modifications for inclusion in a future RCT, enhanced monitoring of that domain within an RCT or to decide that a full RCT would not be feasible at this stage.

Sample size

As this is was a feasibility study, no power calculation was required. By virtue of its status as a rare genetic disorder and acknowledging the multi-faceted impacts on daily

Table 2 Thresholds for evaluation of study feasibility

Outcome	Domain	Progression criteria
Recruitment rate	Over a period of 8 weeks, five participants would be recruited from two sites	Ten participants recruited
Consent rate	Number of eligible people approached against those who consented	≥ 75% people approached consent to study
Adherence	Attendance rate for all sessions in the study	≥ 75% of participants would adhere to ≥ 75% of sessions
Follow-up rates	Completeness of PROMs at each time point	≥ 75% of participants complete PROMs at each time point
Fidelity to the protocol delivery	Delivery of protocol assessed against the following: • Delivery of exercises as described • Delivery of sessions virtually as described	Intervention delivered as described 100% of the time
Safety	Number and type of adverse events and severe adverse events reported by participants	≤ 30% participants would report an adverse event associated with study participation

life living with haemophilia, the potential for research waste in developing and testing novel interventions needs to be avoided. Given the rarity of this condition, coupled with the current contextual difficulties in physiotherapy access for PWH in the UK, the research team decided that 10 participants across 2 sites would be sufficient to allow preliminary evaluation of the feasibility of delivery, data collection and acceptability of this study.

Data analysis

Participant demographics and characteristics were tabulated, and a CONSORT diagram described the flow of participants through the study.

Descriptive statistics were used to assess the feasibility objectives using Excel. Due to the low numbers of participants and in keeping with the feasibility design, continuous variables (outcome measures) were summarised using median, interquartile range and range for group changes between time points.

NVivo (Release 1.6.1 version) was used to manage the qualitative dataset (transcripts and diaries). Acceptability of the intervention was evaluated from analysis of the participant and physiotherapy interviews and diaries.

The interview data and the diary entries from the participants and the physiotherapists were first analysed together using a reflexive thematic analysis approach. This is a six-phase recursive approach comprising the following: (1) familiarisation with the data; (2) coding; (3) generating initial themes; (4) reviewing and developing themes; (5) refining, defining and naming themes; and (6) writing up [47, 48].

A second stage to the data analysis reviewed the completed thematic analysis alongside the initial coding structure developed within. This enabled an analysis of the domains relating to the feasibility and acceptability objectives, helping inform the integration of the qualitative findings with the quantitative data. Quantitative and qualitative results were then tabulated and presented as a joint display.

Results

Study recruitment

Recruitment took place between November 2021 and February 2022 in two large regional haemophilia centres. Twenty-four people were screened for eligibility, 13 were eligible and 10 agreed to take part (consent rate of 77%). Recruitment and retention details are outline in Fig. 1.

Participant characteristics

An overview of participant details is presented in Table 3. Ten male participants aged between 39 and 67 (median age 57) were recruited to the study. Six participants described themselves as independently mobile, three used mobility aids intermittently (cane/crutches) and one used a mobility scooter for longer distances outside. All participants had chronic pain as defined by the International Association for the Study of Pain (IASP) [49] that was present for more than 3 months, and use of pain medication varied.

Primary outcome: feasibility

Feasibility threshold results are presented in Table 4. Target recruitment over an 8-week period was five per site. One site was over-recruited by one participant ($n=6$) and the other site under-recruited by one participant ($n=4$). Adherence rate for the intervention overall was 68.3%. When analysed per session type, adherence rate for the 1:1 session was 84.5% and for the group sessions was 52.1%. There were between-site differences in the attendance rates for the group sessions: Site 01 had 91.7% and Site 02 12.5%.

The reasons given for missing individual 1:1 sessions ($n=8$) included sickness ($n=2$), recovery from an intra-articular ankle joint injection ($n=1$), muscle injury unrelated to the study ($n=1$), joint pain ($n=1$) and knee haemarthrosis unrelated to the study ($n=1$) and nonattendances with no reason given ($n=2$). The reasons stated for nonattendance at the group sessions ($n=25$) were anxiety ($n=6$), other commitments ($n=3$), sickness ($n=1$), flank pain ($n=1$) and no reason given ($n=14$).

All 10 participants (100%) completed baseline PROMs (T0), and 7 (70%) completed at T1 and 6 (60%) at T2. Nine participants agreed to be interviewed at the end of study. One person declined to be interviewed due to anxiety. There were no missing data points identified on any of the outcome measures returned at T1. There were two missing outcome measures for one participant at T2.

Fidelity of the intervention delivery was 84.7%. Sixty-one of the 72 planned individual and group sessions were delivered virtually (as per protocol description) using webcams 80.4% of the time, with the remaining 12 (19.6%) being conducted over the telephone.

Adverse events were recorded. Overall, three participants (30%) reported an adverse event related to the study. Four episodes of increased joint pain after the exercises were reported by three people (one knee, one shoulder, two elbow). One of these participants also reported one episode of hamstring pain the day after the exercise session. Another reported a muscle sprain of his left flank unrelated to the study. One knee joint bleed was reported but was found to be unrelated to the study participation. No serious adverse events were recorded for anyone participating in the study.

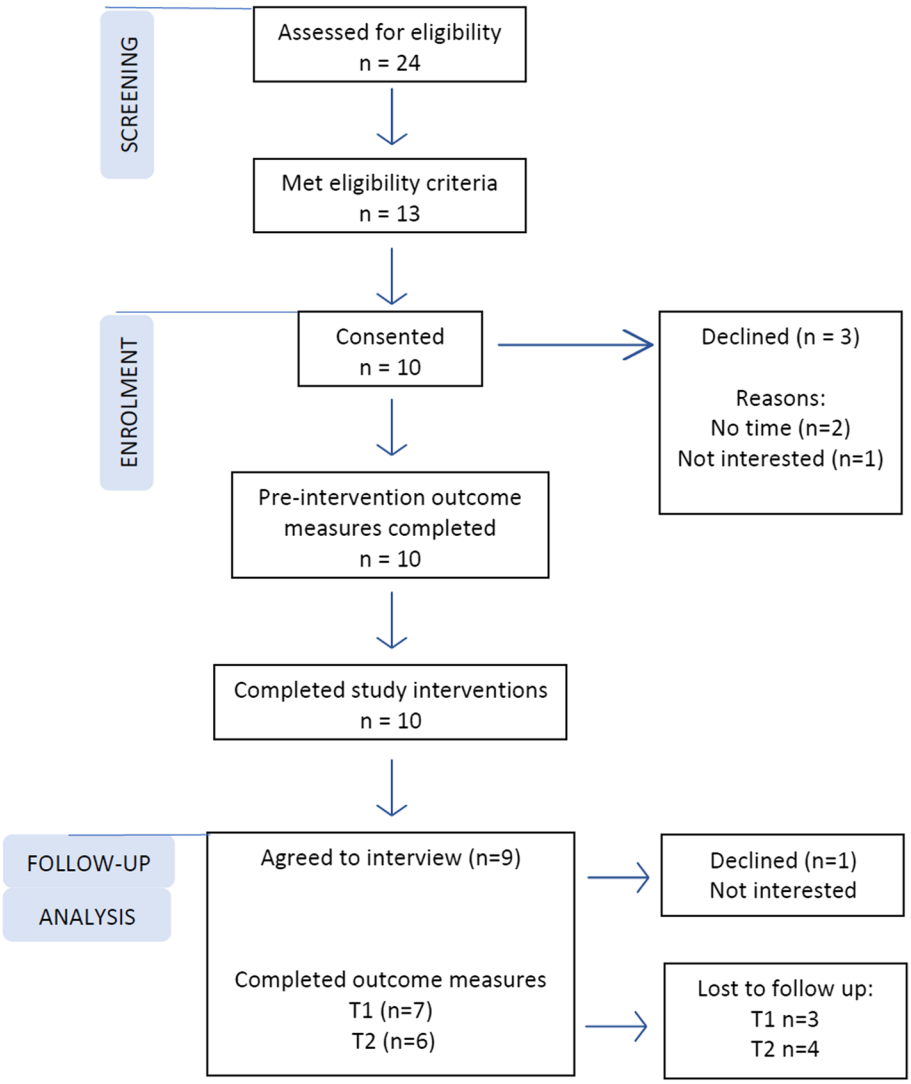

Fig. 1 Participant recruitment and retention in the REMAP-Haemophilia study

The burden of participating in the study was acceptable to participants; however, the burden of study-related administration was highlighted by the physiotherapists. Whilst the organisation and delivery using Microsoft Teams was viewed positively, the time required in the working week to deliver the study as described was deemed significant. The physiotherapists noted that it was just about manageable to host and deliver the sessions, but there were concerns about having time for other tasks such as note writing. Strategies to mitigate against some of these issues included trying to devote a half or whole day to all the appointments or trying to spread them out evenly through the week. The therapists highlighted that 5–6 people would probably be the maximum number of people to include in the study in its current form.

Secondary outcome: efficacy

Group changes in measures of pain, quality of life and function are presented in Table 5 as group median change and interquartile range.

Integrated display of quantitative and qualitative findings

Quantitative and qualitative data for feasibility (Table 6), acceptability (Table 7) and efficacy (Table 8) were collated in a side-by-side format. The level of consensus between the datasets was evaluated as follows:

Table 3 Participant demographics

Variables		*n*	Median (IQR)
Gender — male		10	
Age			57 (21)
BMI			25.95 (2.8)
Diagnosis	Severe Haemophilia A	9	
	- Severe Haemophilia B	1	
Prophylaxis	Trial product	1	
	- Non-factor therapy	3	
	- Standard half-life FVIII	1	
	- Extended half-life FVIII	4	
	- Extended half-life FIX	1	
Employment	Full time	3	
	- Part-time	1	
	- Retired	4	
	- Unemployed	2	
Ethnicity	White British	8	
	- Chinese	1	
	- White, Other	1	
Joints with haemarthropathy	3 or less	2	
	- 4 or more	8	
Comorbidities	HIV	4	
	- Hypertension	3	
	- Liver disease	1	
	- Osteoporosis	1	
	- Peripheral neuropathy	1	
	- Portal hypertension	1	
	- Hypothyroidism	1	
	- Atrial septal defect	1	
	- Previous HCV (cleared)	8	
Pain medications	Acetaminophen	6	
	- COX-II inhibitors	4	
	- Opioids	6	
	- Other — pregabalin	1	
	- Other — *Cannabis*	1	
Orthopaedic surgery	Ankle arthrodesis	4	
	- Knee arthroplasty	3	
	- Hip arthroplasty	4	

- Confirmation — The findings both agree.
- Expansion — The data diverges and expands insights or describes complementary aspects of the topic at hand.
- Discrepancy — The data appear to contradict each other or are inconsistent.

Discussion

This study demonstrates the acceptability, safety and convenience of delivering an exercise-based telerehabilitation intervention for PWH with chronic pain. The protocol was feasible with respect to consent and recruitment rate, adherence to the individual exercise sessions but did not meet the progression criteria for fidelity of delivery and follow-up rates of PROM completion. Both sites recruited successfully to the study. As haemophilia service specifications require people with severe/moderate haemophilia receive biannual clinical reviews [50], it is realistic to assume that recruitment onto definitive trials, and ultimately to the intervention, is achievable. Whilst the chosen PROMs provide little quantitative evidence of change in pain, function or quality of life, the participant interviews did highlight improvements that the participants experienced.

Overall, both study participants and physiotherapists found the virtual delivery acceptable and convenient. However, the physiotherapists reported an increased burden associated with the time needed to deliver the telerehabilitation sessions. Virtual delivery of telerehabilitation has been used in a range conditions such as low back pain, post-operative orthopaedic follow-up and multiple sclerosis, where it has been shown to be comparable to in-person appointments and better than no treatment/intervention at all [51]. However, there remains limited research on the use of telerehabilitation approaches in haemophilia. A study investigating a blended approach (face-to-face physiotherapy and a smart phone application) to rehabilitation for haemophilic arthropathy found this novel approach to be feasible and showed positive effect on lower limb function [29]. Another qualitative study investigated participant experience of a haemophilia-specific exercise class delivered in real time using a smart phone application. They reported similar positive outcomes to this study in respect to convenience and access to clinicians with specialist haemophilia knowledge [28]. Together with the findings of this study, they provide important reflections when considering the use of such technology in day-to-day haemophilia care, particularly when access to specialist physiotherapy remains an issue for up to 60% of PWH [52]. Future studies should include methods to ensure equity of access to digitally delivered healthcare such as telerehabilitation, as well as health economic evaluation to determine cost-effectiveness and how this may be best used to widen access to specialist physiotherapy.

The low-impact, moderate intensity progressive exercise regimen was designed to accommodate participants with multi-joint arthropathy. A Cochrane review investigating exercise interventions and patient beliefs for people with hip and knee OA found interventions are most effective if they are tailored to an individual's preferences, abilities and needs [53]. Whilst it was acceptable overall, some participants said they would have liked a more tailored, joint-specific programme. Future iterations of this

Table 4 Results of feasibility thresholds

Outcome	Domain	Indicator	Result
Recruitment rate	Number of participants recruited over 8 weeks	5 per site	Achieved (partial) — 90%
Consent rate	Number of eligible people approached against those who consented	> 75%	Achieved: 77%
Adherence	Attendance rate for all sessions in the study	> 75%	Partially achieved All sessions = 68.3% Face to face only = 84.5% Group session only = 52.1%
PROM completion	Completeness of PROMs at each time point	> 75%	Not achieved Pre-intervention (T0) = 100% Post-intervention (T1) = 70% 12 weeks post (T2) = 60%

Table 5 Median change in pain, function, and quality of life before and after intervention

		Group median (IQR) at study time points			Median change between time points	
Domain	**Outcome measure**	**T0 (*n* = 10)**	**T1 (*n* = 7)**	**T2 (*n* = 6)**	**T0–T1**	**T0–T2**
Pain	BPI-SF					
	• Worst pain	7 (5)	7 (4)	5 (2)	0	−2
	• Least pain	2 (3)	2 (3)	2 (2)	0	0
	• Average pain	4 (2)	5 (2)	4 (4)	1	0
	• Pain now	3 (4)	3 (4)	3 (4)	0	0
	• Pain interference	5 (5)	3.42 (3.15)	3.28 (3)	−1.58	−1.72
Self-efficacy	PSEQ	45 (27)	39 (27)	37 (20)	−6	−8
HR-QoL	EQ5D-5L					
	• VAS	70 (35)	75 (40)	70 (10)	+5	0
	• Utility score	0.649 (0.308)	0.389 (0.358)	0.698 (0.07)	−0.26	+0.049
	MSK-HQ	30 (14)	39 (14)	35.5 (7)	+9	+5.5
Function	HAL (sum)	46.9 (33)	52.3 (39.1)	49.65 (19.4)	5.4	2.75
	PSFS	3 (1.66)	3 (1)	3.33 (0.84)	0	0.33

study approach will need to consider if including more options for exercise activity based on individual ability is warranted.

The knowledge-sharing and discussion sessions appeared to have limited acceptability. Only one study site managed to conduct the session as described in the protocol, with the other study site being unable to deliver the sessions due to lack of participation with the group sessions. Other studies in haemophilia have included condition-specific education sessions alongside physiotherapy interventions such as manual therapy and exercise, although none has been evaluated for effectiveness within those studies [54–56]. Cochrane reviews evaluating patient education in both RA and OA have shown only small short-term effects for disability associated with RA [57] and no improvements in self-management skills, function or quality of life in OA [58]. This aspect of the REMAP-Haemophilia protocol requires further evaluation and refinement if it is to be included in future studies, in particular how and when PWH want to receive information relevant to their condition.

Evaluating the clinical efficacy of REMAP-Haemophilia was not the primary purpose of this study, but an exploratory analysis on clinical outcomes was included. Although some participants reported some meaningful improvements, these were small, and overall, there were no changes in pain, quality of life and function. Authors have highlighted the need for outcome measures that go beyond just annualised bleed rate and better reflect the improvements in medical care for PWH [59]. A recent publication presented the outcome of a consensus approach to the development a core set of measures to be used in both research and clinical settings in haemophilia. They included number and location of bleeds, health-related quality of life, treatment adherence and joint health [60]. It is clear, however, that a focus on outcomes of disease/condition modification rather than symptom management limits the usefulness of the core

Table 6 Integration of feasibility findings

Topic/domain	Quantitative findings	Qualitative findings	Level of consensus	Key interpretations
Recruitment	10 of 13 agreed to participate: Consent rate = 77% Recruitment rate = 90%	**(Carl, SHA)** — "It was convenience… It seemed like a good idea, the fact I would be having a bit of a session and not having to travel for it." **(Bill, SHA)** — "The biggest thing that piqued my interest … with this it was a little bit more… well, we can hit it [pain and multiple affected joints] from all angles." **(John, SHB)** — "It sounded like an interesting programme and I thought it would be doing something positive as well, kind of giving something back. I didn't know if the exercises… I looked at them and I thought, "Well, I'm not sure if they're going to work or not, I'll give it a go." **(Mark, SHA)** — "I liked the idea of having routine exercises, and I hoped that it would kickstart me into doing something a bit more structured each week."	Confirmation	Progression criteria met Recruitment approaches feasible Virtual delivery and perceived relevance of content positively facilitated decision to take part
Fidelity	Intervention delivery Virtually = 80.4% Telephone = 19.6%	**(Liz, Physiotherapist)** — "There were a couple of people who we did that on the phone rather than having a camera, but that's just… one chap it's just not… that's just not his thing. You just have to take their word for it that they're doing the right thing." **(Bill, SHA)** — "I always did it over the phone. Not because I have a problem with group sessions or anything like that – I was more of a difficult patient because I do shift work. So, the phone calls were easier." **(Mark, SHA)** — "And if the worst comes to the worst you can always switch off the video – you don't have to see people. The important thing is actually hearing the physio going through the programme and timing you and saying, "Three, two, one, start," and then telling you when the 30 s was up." **(John, SHB)** — "There were a couple of times when my Wi-Fi was off on my laptop and I was stuck on my phone, but… Yes, we just had a laugh about that, but that was ok. With modern technology… ok, maybe doing it on a phone isn't ideal but you can still do it."	Expansion	Progression criteria partially met Although fidelity compromised by use of telephone, for those that did use it, it was acceptable in terms of burden and ease of participation Further investigation needed to clarify effect on inclusion and equity in participation

Table 6 (continued)

Topic/domain	Quantitative findings	Qualitative findings	Level of consensus	Key interpretations
Safety	Seven non-serious incidents Increased joint pain ($n=3$) Joint bleed ($n=1$) Muscle pain/sprain ($n=2$) No serious adverse events	**(Jack, SHA)** — "I find them quite safe doing them and not… I mean, even before I was always doing some kind of exercise." **(Luke, SHA)** — "I think I did treat out of prophylaxis once, and that was for… the thigh was really sore. I thought, "Let me treat so that it doesn't mess with the routine. If it is a bleed or if it is something more than just tautness, let me treat this." **(Liam, SHA)** — "For the squats and the lunges I had a chair next to me that I could hold onto… it was there, just a kind of confidence thing, really. So, yes, it felt safe. I had plenty of room around me."	Confirmation	Progression criteria met Study was safe Responsibility to self-assess for additional treatment was not a burden Individualising exercises enhanced feeling safe
Attrition	PROMs completion • T0 = 10 • T1 = 7 • T2 = 6 • Interviews: $N=9$ • No withdrawals		Silence in qualitative data	Progression criteria not met Postal return of PROMs was inconsistent; further studies will need to consider alternate ways to collect data
Adherence	Attendance • All sessions = 68% • Individual sessions = 84.5% • Group sessions = 52.1%		Silence in qualitative data	
Study administration		**(Liz, Physiotherapist)** — "There was a lot of… obviously, the admin side of it, but a lot of that is just because it's a study. Because I was thinking, practically, how could this work if you do it outside of a study." **(Liz, Physiotherapist)** — "I had them each hour and that was enough time for me to then do the little bit of a start, do the main session, say goodbye, do the notes, do the next one … you needed that hour to do the admin bits either side. Yes, 45 min would have been stressful to do all five in a one till five chunk of time." **(Dan, Physiotherapist)** — "It does take up quite a lot of time doing it that way… I got quite lucky with the participants, that once I had them in and we just said, "Ok, well, this is what we're going to do and these are the proposed dates. How does that work for you?" there was really hardly any issues with all six of them." **(Dan, Physiotherapist)** — "Six would be a… six would be, I would say, an absolute max. So, it would be doable with enough planning, but I think it would be a challenge. Because it essentially takes up a day of work when you've got the six."	Silence in quantitative data	Whilst virtual delivery viewed positively, perceived increased burden of study administration by physiotherapists Further studies will need to be mindful of time required to deliver study against physiotherapist hours available for haemophilia services

Table 7 Integration of acceptability findings

Topic/domain	Quantitative findings	Qualitative findings	Level of consensus	Key interpretations
Protocol — virtual delivery		**(Liam, SHA)** — "I mean, it works and it doesn't impinge on your day, as it were. If I was coming up to you, it's an hour to you and an hour back again, so that would be a morning gone. Whereas doing it this way, it's an hour roughly, 45 min or whatever, and then you get back and you're at home and you get on with doing what you want to do. And that really worked, I thought." **(Jack, SHA)** — "I think in the past couple of years we're doing the same thing for everything. We're in front of a screen now and you just become… like, again, more normalised. It's not much different, really, I suppose." **(Luke, SHA)** — "I suppose turn on the camera, do your thing, 40 min later you go and get on with the rest of your day – not two hours later you get on with the rest of your day, or two and a half hours later you can get on with the rest of the day. I mean, even for me to get to the Royal Free, it's 45 min or something – get to the station… or if I drive, even… So, that's an hour and a half each way, maybe. This is obviously more convenient." **(Adam, SHA)** — "face-to-face is better, in that you get much… Certainly the personal face-to-face session, you really… You can't convey the full physicality of what you're doing on a Teams call."	Silence in quantitative data	Virtual delivery acceptable, it although remained a desire for option of face to face in combination

Table 7 (continued)

Topic/domain	Quantitative findings	Qualitative findings	Level of consensus	Key interpretations
Protocol — exercises		**(Liz, Physiotherapist)** — "But I think just that general conditioning and general fitness was something that was quite… it was within everybody. I think so many people who we see have not been able to do what they want to do because of pain or for whatever reason haven't pushed themselves. So, I think it fits when it's very generalised." **(Carl, SHA)** — "We had tailored it more to just to some… They were still doing a couple of the other movements because… you know, it's good to get you panting and your heart rate up, so we were doing some of the other ones which was good as well. But by the end of it we were doing more targeted stuff for my… you know, for my ankle." **(Adam, SHA)** — "I think six weeks is just about right. It allows people to learn as they go along. It allows for the odd session that you have to miss. So, it's a good length of time. I'm happy enough with that." **(Mark, SHA)** — "Thirty second rest between each… I'm not sure what the right… set, between each set. The two minutes between each exercise, each set of exercises, I felt was probably a bit long. You could probably get away with a minute, or a minute and a half. Some of us were sort of twiddling our thumbs a bit for two minutes."	Silence in quantitative data	Exercises were acceptable, particularly because it permitted alterations to fit ability As weeks progressed, some identified that they would have preferred a more specific single joint programme as well

Table 7 (continued)

Topic/domain	Quantitative findings	Qualitative findings	Level of consensus	Key interpretations
Protocol — knowledge and discussion sessions	Only one site completed the knowledge and discussion sessions as described	**(Hugh, SHA)** — "It didn't… well, it didn't actually happen, really. I think there were technical issues and also when I was going to join them I think… well, basically no one else was left." **(Liam, SHA)** — "The slides were interesting as well because it just kind of gives you a reminder of what you're doing and what to look out for and that kind of thing. And knowing your limits and then perhaps after a few weeks just trying to push those limits" **(Luke, SHA)** — "I can't dismiss it because I'm sure that there were moments in each PowerPoint where it was like 'yes'. And even if that's a personal back of the head 'yes' and you're identifying with something, so you're feeling this person gets it, that's very valuable. I don't think any of it was revelatory." **(Hugh, SHA)** – "I still struggle with the pacing because if it feels good then I think, "Well, I'm ok now." But I'm starting to realise now that it's not necessarily the case. But the pacing was good. And understanding different types of pain was definitely good"	Discrepancy	Unclear if knowledge and discussion sessions in current form are acceptable due to only one group completing them The sessions did not enhance the experience of taking part

Table 7 (continued)

Topic/domain	Quantitative findings	Qualitative findings	Level of consensus	Key interpretations
Outcome measures used		**(Carl, SHA)** – "I mean, a lot of those surveys do feel, you know, quite kind of generic. And you know, on a scale of 1 to 10… I bet they differ every time I fill them out. You know, I think about that every time when I'm filling them out and go, "God, what did I put last time and is it different?" **(Luke, SHA)** – "Some of them I had to answer… like, "How did you feel in the last 24 h?" "I felt shit. I wasn't able to walk," kind of thing. Whereas that wasn't really a picture of the last three months. It does squeeze you into answering in a particular way which might not be relevant on the whole." **(Adam, SHA)** – "They weren't precise enough. I mean, they were sort of flopsy questions, really."	Silence in quantitative data	Time to complete PROMs was acceptable PROMs had limited value to participants, reported as lacking specificity to them, their condition and their experience
Enjoyment		**(Dan, Physiotherapist)**—"I think it was… like, from the start, from quite early, it seemed like there was… it felt like a positive thing. So, it felt enjoyable for me and I got the sense from the participants that they were enjoying it and benefiting from it as well. So, it made it an enjoyable experience." **(Luke, SHA)** – "It was very useful, it was very beneficial. I could see a need for it, I could see a desire for it, even if the person is not necessarily aware of the desire for it. I felt good doing it. I'm glad you did it and I'm glad I was able to be part of it." **(Jack, SHA)** – "I felt really relaxed. I didn't have to think about… concern about anything. But in a different situation with different people, then you wouldn't be able… you always think about "I'm different to them". And definitely, it makes you just relax a lot more and just get on. Definitely you feel more enjoyable doing it."	Silence in quantitative data	High degree of acceptability was associated with high level of enjoyment Seeing others like them, and exercises individualised to them promoted enjoyment

Table 7 (continued)

Topic/domain	Quantitative findings	Qualitative findings	Level of consensus	Key interpretations
Motivation		**(Hugh, SHA)** – "it was good to take part and it's helped me, I think, get more confident and get out of… I think there was a little rut and I think that should help me stay out of it and get a bit more consistent with some of these exercises. So, it was good to take part, yes." **(John, SHB)** – "I think my plan is that… Because I've got an exercise bike that I kind of go on, so I thought if I did alternate days, one day on the bike and one day running through those exercises – that would be the plan, anyway – I'd do that."	Silence in quantitative data	Positive influence on motivation with increased confidence to do more and make plans for further exercise activity

Table 8 Integration of efficacy findings

Topic/domain	Quantitative findings	Qualitative findings	Level of consensus	Key interpretations
Pain	BPI domains *Worst pain, least pain, pain now* • No change in group median *Pain interference* • Group median decrease of 1.58 points PSEQ • Group median decrease of 6 points	**(Adam, SHA)** — "What I mean by that is it's not so much the pain is necessarily less, it's the confidence with which you can cope with it has improved." **(Bill, SHA)** — "I've got… It's just less pain. All the pain is less, if that makes sense. The pain is a little less. It's still there but… Like, for example, bending … that kind of movement is now not sparking as many issues." **(Adam, SHA)** — "you kind of… it sort of carries the pain. I don't know how to put it. It's like the pain becomes a passenger as opposed to a driver. It's not the dominant thing that I'm thinking about now." **(Carl, SHA)** — "It's always maintained the same, my pain is consistent. So whether I felt any extra pain after doing the exercises – no more than I normally would from doing those kind of exercises, which I've done my whole life. So there was nothing… no difference in pain to report over the six weeks period"	Expansion	PROMs suggest limited effect of intervention; however, qualitative data highlight positive individual changes on day-to-day activity despite pain Highlights value and importance of evaluating the personal experience of interventions targeting pain
Function	HAL • Small increase group median of 5.4 points PSFS • No change	**(Hugh, SHA)** — "It does make you realise that it's one of those things where the strength of your body will help with the arthritis. I think that's something I didn't really consider that much, whereas now I'm a bit more aware of it." **(Mark, SHA)** — "Well, it was just showing me that actually I… I find certain things difficult to do and I realised that they all find them difficult to do as well, so it's not just me being particularly bad." **(Adam, SHA)** — "There's still a bit of pain going upstairs – the most tangible thing that's improved is going up and down stairs with both legs as opposed to one… using my left leg only and my right leg, with the weak quads and things, the one where I had the knee replacement… So, I just feel more in control of it. And your balance is a little bit better."	Expansion	Appears to be a functional improvement which is only noticed after taking part Identifying goals upfront may not be effective if people are unaware what has potential to change

Table 8 (continued)

Topic/domain	Quantitative findings	Qualitative findings	Level of consensus	Key interpretations
Quality of life	EQ5D5L — VAS • Increase in group median of 5 points EQ5D5L — Utility • Decrease in group median = 0.26 MSK-HQ • Group median increase = 9 points	**(Adam, SHA)** — "It really opened my eyes to how important it is, how quickly it can make a difference to your body. And I just wish I'd been doing this much earlier." **(Luke, SHA)** — "This really makes you feel like someone's listening and someone's caring enough to do something –that's another hidden benefit, is that you're feeling part of… you and the unit are feeling part of the same thing. Which is great." **(Liam, SHA)** — "I feel better… I mean, for exercising. And I think the process of doing the exercising is good mentally and afterwards you feel "Yeah, I've done it."	Discrepancy	Minimal change in QoL measures — however, qualitative findings indicate individual positive changes in mood and seeing/feeling physical changes
Subjective view of improvement	Patient global impression of change ($n = 7$) • Worse ($n = 1$) • No/minimum change ($n = 4$) • Much improved ($n = 2$)	**(Luke, SHA)** — "It was very useful, it was very beneficial. I could see a need for it, I could see a desire for it, even if the person is not necessarily aware of the desire for it. I felt good doing it. I think it would be useful to me." **(John, SHB)** — "And there's no doubt that if you're doing structured exercises they have a positive impact on your wellbeing and your health because you get more adept at doing them, then you can have more of an exertion because you're putting more into them, and you do… it does uplift you that kind of physical activity" **(Adam, SHA)** — "I think I do feel a bit bouncier. And that has effects on mood as well as your general… the mobility that goes with greater fitness. It's not that it's affecting the target joints particularly; it's more that it's affecting everything around it."	Discrepancy	

Table 8 (continued)

Topic/domain	Quantitative findings	Qualitative findings	Level of consensus	Key interpretations
Behavioural modifications		**(Liam, SHA)** — "Yes, it's probably given me a bit more impetus, again, to do more now I've been shown… you know, doing exercises at home and ways of doing it and different types of exercises." **(Carl, SHA)** — "To be honest with you, I could be doing more." And she said, "Right, ok, cool." And so I said, "So, you're all right if I just do more… obviously not pushing it or going to crazy levels, but just… If you're in a position where you can do your exercises…" So, I did it every other day." **(Jack, SHA)** — "at the moment, yes, I still have that enthusiasm and want to do it and get on with it and do something about it. But in a month, two months later… I can't say anything." **(John, SHB)** — "Since the programme finished – it was only a week or so ago, a couple of weeks ago – I've tried doing some of those exercises on my own because I think they're worth carrying on with."	Silence in quantitative data	Inclusion of BCTs to encourage participation was successful — some participants describe a desire to change or actively implementing changes to change

set for pain research in haemophilia. Whilst the participants accepted the need to collect measures, they were less accepting the applicability of the PROMs. This may be because the assessments did not fully encompass their individual view of their haemophilia and pain experience, reflected in the interviews findings (Table 6) describing 'generic' surveys and feeling 'squeezed into answering in a particular way'. More work is required to establish an acceptable method of measuring impact of rehabilitation interventions in PWH and perhaps giving more consideration/weight to the overall qualitative experience.

A strength of this study was the application of mixed methods in the data collection and analysis, the inclusive approach to recruitment and the pragmatic protocol design to encourage and facilitate participation in the study activities. A mixed-methods approach enables consideration of multiple viewpoints and positions to achieve a deeper understanding of the study findings [61]. Understanding the experience of all involved in the delivery and participation of the study, as well more practical issues concerning burden of delivery and administration, means this study adds to the current quantitatively heavy evidence base of physiotherapy rehabilitation in haemophilia. A follow-up review and evaluation of the programme theory that underpins this study [33] are planned to identify changes and further refine the protocol for a more definitive future trial.

The inclusion criteria for this study were purposefully broad, reflecting real-world experience and acknowledging the highly complex nature of PWH living with multiple joint arthropathy and chronic pain. When working with people with rare disorders such as haemophilia, it is important that study design does not further marginalise those who may have most to gain from taking part. This is especially important for PWH as it remains unclear if established rehabilitation programmes addressing predominantly single joint issues such as ESCAPE-pain [62] and the GLA:D OA knee [63] would be suitable.

Another strength is the inclusion of two sites in this feasibility study. This meant the study was able to include two different groups of PWH under the care of different specialist physiotherapists. This was an important consideration for feasibility. If there were difficulties delivering this study at a local level within well-staffed haemophilia centres, then it is highly likely that it would not be at all feasible in centres with less than full-time physiotherapy input.

The main limitation of the study is the small number of participants, so no statistical inferences can be made. The study's feasibility design aimed to see if the intervention could be delivered and if it was safe and acceptable [64]. Poor completion rates of the post-intervention PROMs at each time point impaired preliminary evaluation of efficacy of the intervention. Further work is required to ascertain what PROM's participants consider valuable to them (e.g. generic or condition specific), when and how to collect them and the most effective way of collecting data such as digital/electronic forms.

Another limitation is that whilst almost all participants had more than three joints affected by haemophilic arthropathy, the whole-body approach of the exercise programme was not acceptable to all participants, with some wanting a more joint-specific exercise approach. Future studies will need to balance the practicalities of a high degree of individualisation, alongside evaluating feasibility and applicability of an exercise programme in a population-based representative cohort such as those here.

Conclusion

The present study demonstrated an exercise-based, telerehabilitation intervention for people with haemophilia is feasible, safe and acceptable. Further work is needed to evaluate the choice of objective outcomes used and how they are collected, as well as the value of including a more subjective, person-centric experience of taking part in studies such as these.

Abbreviations

PWH	Person/people with haemophilia
MRC	Medical Research Council
REMAP-Haemophilia	REhabilitation for the Management of Arthritic Pain in Haemophilia
PROM	Participant-reported outcome measure
BCT	Behaviour change technique

Supplementary Information

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Supplementary Material 1. List of Behaviour Change Techniques included in design and delivery of the REMAP-Haemophilia feasibility study.

Supplementary Material 2. Exercise description and level of difficulty for the REMAP-Haemophilia study.

Supplementary Material 3. REMAP-Haemophilia Knowledge and Discussion session presentation slides.

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Authors' contributions

All authors contributed to the overall study design. PML was the chief investigator for this study. All authors contributed to the conceptualisation and development of the study, protocol, ethics application and document preparation. All authors reviewed, edited and approved the final version of this manuscript.

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Availability of data and materials
The datasets used in this study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate
The study was conducted in accordance to the ethical approval granted by the East Midland-Nottingham 2 Research Ethics Committee (Rec. reference number: 21/EMI/0161).

Consent for publication
Not applicable.

Competing interests
The authors declare that they have no competing interests.

Author details
[1]Katharine Dormandy Haemophilia Centre and Thrombosis Unit, Royal Free London NHS Foundation Trust, London, UK. [2]Department of Academic Haematology, University College London, London, UK. [3]Haemnet, London, UK. [4]The Haemophilia Society, London, UK. [5]School of Medicine, Health and Social Care, Canterbury Christ Church University, Canterbury, Kent, UK. [6]St. George's University of London, London, UK.

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